The importance of venous hypertension in the formation of dual arteriovenous fistulas: a case report of multiple fistulas remote from sinus thrombosis

Abstract Various hypotheses have been reported concerning the pathogenesis of dual arteriovenous fistulas (DAVF). However, it is still controversial whether sinus thrombosis or venous hypertension has a greater influence on the formation of DAVFs. We present a rare case of multiple DAVFs that developed after sinus thrombosis. Chronic venous hypertension secondary to sinus thrombosis in the left transverse-sigmoid sinus induced the multiple DAVFs, including one in the right cavernous sinus, which was remote from the occluded sinus. This case indicates the importance of venous hypertension in the formation of DAVFs.

Keywords Multiple dual arteriovenous fistulas · Venous hypertension · Sinus thrombosis

Introduction
Dural arteriovenous fistulas (DAVFs) comprise 10–15% of intracranial arteriovenous malformations [1]. They most commonly involve the transverse, cavernous and sigmoid sinuses [2]. Various hypotheses have been developed concerning the pathogenesis of DAVFs and whether they are congenital or acquired. In recent years, DAVFs have been generally considered to be acquired lesions [3–12]; however, whether sinus thrombosis or venous hypertension has a greater influence on their formation remains controversial.

We present a rare case of multiple DAVFs that developed in a patient after sinus thrombosis. The right cavernous sinus DAVF developed remotely from the thrombosis. These findings suggest the importance of venous hypertension in the formation of DAVFs.

Case report
A 55-year-old man presented with a sudden onset of severe headache and disturbed consciousness, and was admitted to another hospital in March 1996. A computed tomography scan showed intraventricular hemorrhage (Fig. 1a). Magnetic resonance imaging showed a fresh thrombus in the left transverse-sigmoid sinus, which was revealed as iso-intensity on the T1-weighted image (Fig. 1b) and high intensity on the T2-weighted image (Fig. 1c). Initial angiography of the left common carotid showed no abnormal findings on the arterial phase (Fig. 2a) but did show occlusion of the left transverse-sigmoid sinus on the venous phase with contrast filling of the right cavernous sinus (Fig. 2b). Angiography of the right common carotid also showed no abnormal findings on the arterial phase and no sign of thrombosis in the cavernous sinus. The patient was treated conservatively, and had complete resolution of the symptoms.

Nine months later, the patient presented with right conjunctival chemosis and exophthalmos, though a bruit was not heard. Angiography performed 10 months after the first admission showed multiple DAVFs involving the left transverse-sigmoid sinus and the right cavernous sinus. The patient was referred to our institute for further treatment in February 1997. Angiography of the left external carotid revealed the left transverse-sigmoid sinus DAVF, which was supplied by bilateral occipital arteries, and the left superficial temporal, middle meningeal, posterior auricular, and ascending pharyngeal arteries. The lesion drained retrogradely into the right transverse sinus through the left transverse sinus (Fig. 3a, b). Angiography of the right carotid revealed the right cavernous sinus DAVF, which was remote from the occluded sinus (Fig. 4a). This DAVF drained into the right superior ophthalmic vein.

On February 25, transvenous embolization was done through the right inferior petrosal sinus via the right jugular vein. The cavernous sinus DAVF was then occluded completely with platinum coils (Fig. 4b), and the patient’s ocular symptoms disappeared.
Fig. 1  a On admission a CT scan shows intraventricular hemorrhage. b, c MRI showing a fresh thrombus in the left transverse-sigmoid sinus, which was revealed as iso-intensity on the T1-weighted image (single arrow) (b) and as high intensity on the T2-weighted image (double arrows) (c).

Fig. 2a, b Left common carotid angiograms. a Lateral views, showing no abnormal findings in the arterial phase. b Venous phase, anteroposterior views, showing that the left transverse-sigmoid sinus was occluded (arrow) with contrast filling of the right cavernous sinus.

within several weeks. On March 11, the feeding arteries of the left transverse-sigmoid sinus DAVF were embolized through a transarterial approach with polyvinyl alcohol (PVA) particles. This treatment markedly reduced the arterial flow into the transverse-sigmoid sinus.

Six months later, the patient reported having left-side pulsatile tinnitus, and follow-up angiography revealed no contrast filling of the right cavernous sinus DAVF but an enlarged left transverse-sigmoid sinus. In October 1997, transarterial embolization of the DAVF was performed again with PVA particles and platinum coils. After this treatment, the tinnitus disappeared.

In August 1999, the patient again presented with left-side tinnitus, and follow-up angiography revealed the recurrence of the left transverse-sigmoid sinus DAVF (Fig. 5a). On December 22, transvenous embolization was done through the right transverse-sigmoid sinus via the right jugular vein. Before embolization, the mean pressures in the superior sagittal and right transverse-sigmoid sinus were 20 mmHg, which indicated chronic venous hypertension. The tip of the microcatheter was then advanced into the left transverse-sigmoid sinus close to the fistula. At that time, we felt some resistance from what seemed to be the organized thrombus. The mean pressure in the left transverse-sigmoid sinus was 77 mmHg by arterial inflow. The affected sinus was embolized with platinum coils. Left common carotid angiography performed after embolization showed that the DAVF has been obliterated (Fig. 5b). After this treatment, the sinus pressure returned to normal. The mean pressure in the left transverse-sigmoid sinus was 11 mmHg, and in the superior sagittal and right transverse-sigmoid sinuses was 6 mmHg. The tinnitus disappeared after embolization, and the patient has done well, and has remained free of any further incidents for 6 months.

Discussion
Dural arteriovenous fistulas constitute 10 to 15% of intracranial arteriovenous malformations [1]. They most commonly involve the transverse, cavernous and sigmoid sinuses [2]. Multiple DAVFs are rare [5, 7, 12–25]; the prevalence has been reported to be 7%